Arteriovenous Fistula in a Nerve Root of the Cauda Equina Fed by a Proximal Radiculo-Medullary Artery

A Report of Two Cases

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Summary

While there have been a few reports on cases of intradural spinal arteriovenous fistula located on the filum terminale, no cases of its location in a nerve root of the cauda equina have been reported to date.

We describe two such cases and describe the intraoperative findings. A 40-year-old man presented weakness of his left leg. Another 62-year-old man presented paraparesis dominantly in his left leg with urinary hesitation. In both cases, spinal T2-weighted magnetic resonance images showed edema of the spinal cord, indicating a flow void around it. Digital subtraction angiography disclosed an anterior radicular artery branching from the anterior spinal artery on the surface of the conus medullaris and a turnaround vein running in the opposite direction within the cauda equina.

In the first patient, while the feeding artery running along a nerve root was detected, the draining vein and the fistula were not identified at first sight. An incision into the respective nerve root exposed their location within it. In the second patient, unlike the first case, the feeding artery and the fistula were buried in a nerve root, while the draining vein was running along the nerve's surface. In both cases, permanent clips were applied to the draining vein closest to the fistula. The recognition of a hidden fistulous point in a nerve root of the cauda equina is essential for successful obliteration of the fistula.

Introduction

With regard to intradural spinal arteriovenous fistulas (AVFs) in the level of the conus medullaris, it seems that the artery arising from the continuity of the anterior spinal artery (ASA) beyond the tip of cauda equina has been thought to pass along the filum terminale and that the fistula to be formed thereon. There have been few reports on such cases identified by direct surgery in the past [1-5].

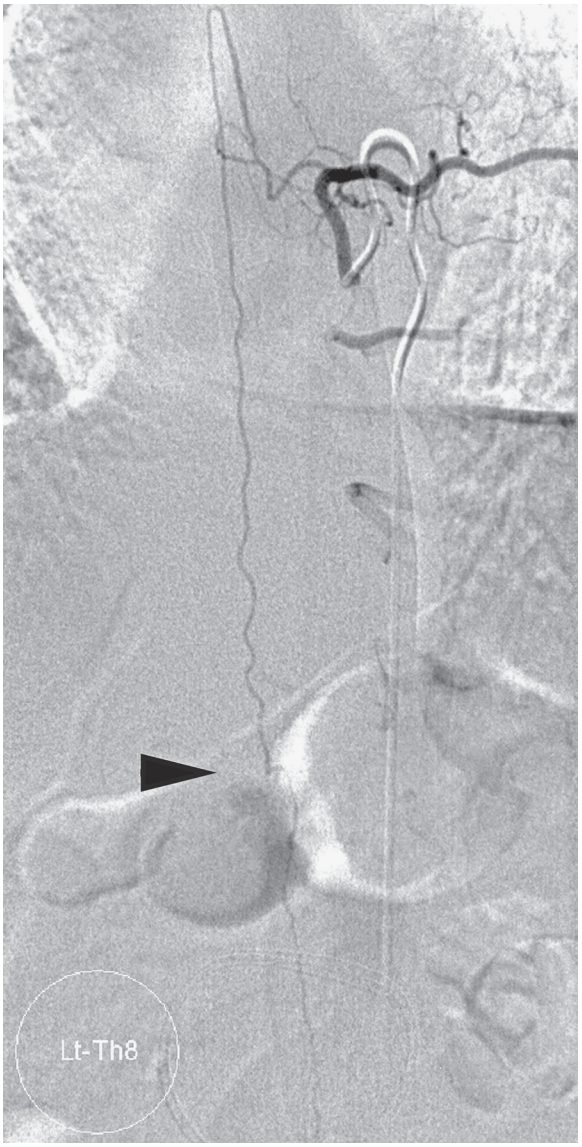
However, cases of the fistula being located into a nerve root of the cauda equina have not been reported to date. Within the cauda equina, angiographical analyses cannot detect the exact anatomical point of the fistula, even though they might have suggested its level or the angioarchitecture.

This report describes two cases of fistula in a nerve root of the cauda equina fed by a proximal radiculo-medullary artery and details the intraoperative findings.

Case Report

Case 1

A 40-year-old man presented sudden weakness of his left leg with paresthesia. Spinal T2-weighted magnetic resonance (MR) imaging showed a slight swelling of the conus medullaris and some low intensity signals around it indicating a flow void (Figure 1A).



Digital subtraction angiography (DSA) disclosed the artery of Adamkiewicz by the left T-8 intercostal artery angiogram, revealing an anterior radicular artery running in a caudal direction branching from the ASA on the surface of the conus medullaris (Figure 1B).

The fistula was identified between the artery and a turnaround vein running in the opposite direction (Figure 1C).

In the operation, the patient was placed in the prone position, and the lower lamina of L-1 and the complete lamina of L-2 were removed bilaterally. While a midline durotomy exposed the feeding artery running along a nerve root of the cauda equina, the draining vein and the fistula were not identified at first sight. An incision into the respective nerve exposed their location within it. A permanent clip was applied to the closest draining vein on the distal side of the fistula, within the nerve root (Figure 2A).

The weakness of the patient's left leg was immediately resolved after the operation.

Spinal angiography performed one week after surgery showed a complete obliteration of the fistula. Follow-up MR imaging eight months after surgery demonstrated that there was no recurrent AVF (Figure 2B).

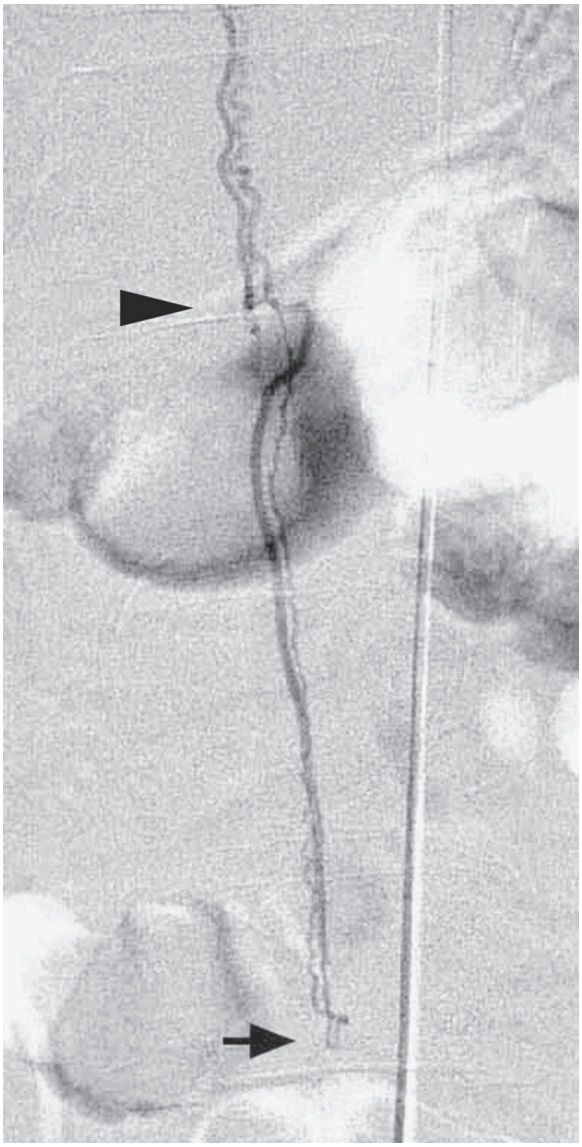


Figure 1 A) Preoperative sagittal T2-weighted magnetic resonance imaging scan showing slight swelling of the conus medullaris and flow void of the surrounding tortuous veins. B) Angiograms. An anterior radicular artery branched from the anterior spinal artery is seen (arrow head). C) Arrow indicates the location of fistula. Note the ascending serpentine perimedullary vein. Arrowhead corresponds to that B.

Case 2

A 62-year-old man had observed the symptoms of left leg hypesthesia, paraparesis dominantly in left leg, and urinary hesitation for three weeks before the treatment. Spinal T2-weighted MR imaging showed marked edema of a wide area of the thoracolumbar spinal cord, indicating flow void on its ventral and dorsal side (Figure 3A). The DSA findings seemed to be quite similar to Case 1 (Figures 3B and 3C). The intraoperative findings, however, revealed opposite features on the angioarchitecture as compared to *Case 1*. Unlike *Case 1*, the feeding artery and the fistula were completely buried in a nerve root, while the draining vein was running along the nerve's surface. Therefore, a permanent clip was applied to the surface draining vein most proximal to the fistula (Figure 4A). Postoperatively, the patient's paraparesis and urinary dysfunction gradually improved, and he became able to walk without assistance. A spinal angiogram obtained two months after the surgery confirmed the obliteration of the AVF. Although the spinal cord edema remained to a certain degree according to the T2-weighted MR imaging conducted eight months after surgery,

Figure 2 A) Intraoperative photograph showing the clipped draining vein which is buried in a nerve root of the cauda equina. B) Postoperative sagittal T2-weighted magnetic resonance scan obtained 8 months after surgery, revealing disappearance of flow voids around the conus medullaris.

the flow void signals disappeared almost completely (Figure 4B).

Discussion

Intradural spinal AVF was first reported by Djindjian et al. in 1977. In 1987 Gueguen et al. advocated classifying intradural spinal AVFs into three types [3]. Type 1 lesions have a simple and small extramedullary fistula fed by a single artery. Type 2 lesions consist of a medium-sized fistula fed by one or two feeding arteries which drain into a dilated vein. Type 3 lesions have giant fistulas fed by arteries with wider diameters, causing rapid venous drainage or large dilated varices. These intradural spinal AVF types were subsequently reclassified by Spetzler et al.

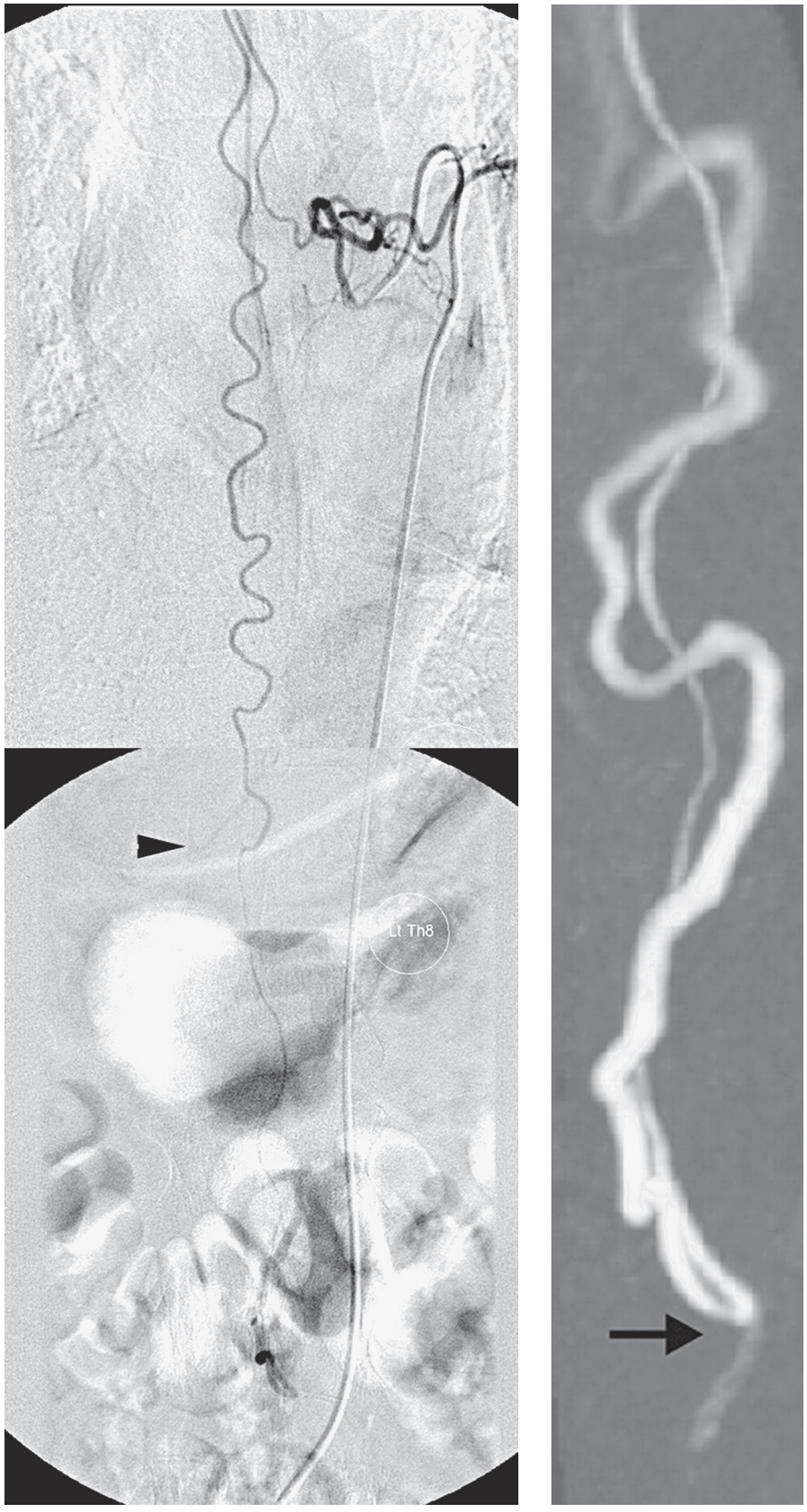


Figure 3 A) Preoperative sagittal T2-weighted magnetic resonance imaging scan revealing intramedullary hyperintensity signals of the thoracolumbar spinal cord, as well as serpentine dilated perimedullary veins. B) Digital subtraction angiography (DSA) showing an anterior radicular artery branched from the anterior spinal artery (arrowhead). C) 3D DSA showing the angioarchitecture of the anterior radicular artery as the feeding artery, the fistula (arrow), and ascending vein in a cephalad direction.

A

B

Figure 4 A) Intraoperative photograph showing the clipped draining vein running along the surface of a nerve root of the cauda equina, in which the feeding artery is buried. B) Postoperative T2-weighted magnetic resonance imaging scan obtained eight months after surgery, showing some residual spinal cord edema but also a reduction of flow void signals.

into ventral and dorsal subtype groups. The first group consists of subtypes A, B, C, whose angioarchitectural characteristics correspond to those of Gueguen et al., while the second group distinguishes between the two subtypes A (with a single feeding artery) and B (with multiple feeding arteries) [6,7]. However, the classification of intradural spinal AVFs is still controversial for there to be any disagreement.

In angiographical findings, it seems to be relatively difficult to detect the precise anatomic fistulous point. However, a few studies give the resolutions to this obscurity. Parke et al. [8] and Crock and Yamagishi [9] presented a detailed anatomy of the conus medullaris and cauda equina blood supply by vascular injection. Furthermore, they showed how the radicular arteries distribute their intraradicular branches to the individual nerve root filaments to be a multiple "step-down" process. Berenstein and Lasjaunias showed the detailed angioarchitecture of the total spinal cord and the cauda equina, including the clinical diagnosis and the treatment of the spinal AVFs [10,11].

Regarding the two cases presented in this paper, the feeding artery seems to be an anterior radicular artery branching from the ASA before the basket on the anterior surface of the conus medullaris. Other collateral arteries might have vascularized the shunt, but such

feeding arteries were not detected by selective spinal angiography as far as the L3 artery at the caudal side or by multidetector-row computed tomographic angiography. Therefore, this is the first report of two cases in which the fistula was located in a nerve root of the cauda equina. Since embolization of the ASA or their direct branches may incur irreversible complications, direct surgery tends to be favored recently as the treatment of ventral Type 1 lesions [4,12]. Similarly, it is important to recognize such lesions, having the fistula in a nerve root of the cauda equina, fed by the artery branching from the ASA and possibly the posterior spinal artery or the basket.

Conclusion

The recognition of a hidden fistulous point fed by the anterior radicular artery branching from the ASA, into a nerve root within the cauda equina is essential for successful obliteration of the fistula.

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